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Clinical case

Primary cervical malignant teratoma with a rib metastasis in an adult: Five-year survival after surgery and chemotherapy

A case report with a review of the literature

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Summary

We report a case of a man presenting with a cervical malignant teratoma and a chondrosarcomatous rib metastasis. He was alive and free of recurrence five years and 10 months (= 70 months) after resection of the primary mass, followed by chemotherapy and subsequent resection of the rib tumor. This is the 35th patient reported in the literature and the first description in which an 'adjuvant' or primary chemotherapy was used. Previous patients with a cervical malignant teratoma, reported after lethal outcome, had survivals of one to 22 months (median nine months). In all patients with a preoperative clinical impression of an aggressive, differentiated or undifferentiated malignancy, the definite diagnosis of teratoma could only be made histologically. By analogy to germ cell tumors, the prognosis of malignant teratoma might be improved if complete excision is combined with new, adjuvant chemotherapy protocols for germ cell tumors. Lessons learned from this case are placed in the context of germ cell tumors in general and of non-gonadal malignant teratomas in particular.

Key words: chemotherapy, cervical, germ cell tumor, prognosis, surgery, teratoma

Case report

a) History and clinical findings

A white, 33-year-old man was referred because of a mass on the anterior left side of the neck. He had first felt the painless nodule a month previously and was aware that it had grown after this date. There were no symptoms of mechanical obstruction. He had no previous history of any operation of or irradiation to his neck. On *clinical examination*, he was in a good general condition, without any distress. A firm, elastic, 6 × 5 cm tumor mass was situated in front of the medial rim of the left sternocleidomastoid muscle and extended into the left thoracic aperture. It was fixed to the underlying structures but not to the skin. There were no palpable lymph nodes. *Laryngoscopy* was normal.

b) Preoperative investigations

A chest X-ray showed a focal swelling of the 10th left rib due to a poorly delineated osteolytic lesion, compatible with an enchondroma. A *computerized tomography of the neck* (Figure 1) after contrast injection showed an inhomogeneous tumor mass (8 × 4 cm). This was lateral to the thyroid with compression of the left thyroid lobe, of the left side of the trachea and the left neck vessels and with possible infiltration of the jugular vein. The density of the mass was 11–73 HU. Routine blood and urine examinations were normal. Thyroid studies revealed euthyroidism with normal values of thyroxine, triiodothyronine, thyrotropine. The *thyroid uptake* (two hours, 123-I, 7.4 MBq) was 9%. The 1 : 1 *thyroid scan* (123-I and 99mTc-Methoxyisobutylisonitrile [4] (MIBI, Cardiolite®

Figure 1. Transverse CT section of the neck after intravenous injection of contrast media: note the large, inhomogeneous tumor mass in a left supraclavicular location, measuring 8 × 4 cm in diameter. The tumor density was 11–73 HU. Infiltration of the left internal jugular vein was suspected. Compression of the left tracheal wall, displacement of the left thyroid lobe, of the left vessels and of the sternocleidomastoid muscle were noted.

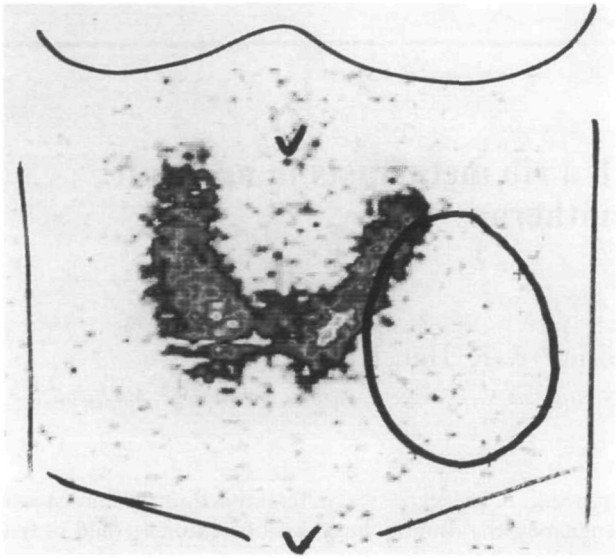

Figure 2. Thyroid scan, performed two hours after oral administration of 7.4 MBq 123-I (Polyscan), showing a homogeneous activity distribution within a slim, 20 g gland. The lateral margin of the left lobe is compressed by a cold nodule of 6 × 5 cm diameter.

Dupont, USA): 185 MBq) showed homogeneous activity distribution in a slim, 20 g gland (Figure 2). The lateral margin of the left lobe was impressed by a cold nodule of 6 × 5 cm diameter. Two-dimensional *ultrasonography* showed a well delineated nodule of inhomogeneous echostructure lateral to and compressing the left thyroid lobe. Several small cystic areas, a few hyperdense foci, but no calcium deposits were noted. *Fine needle aspiration biopsy* produced only cystic fluid without cellular elements suspicious of malignancy. Cytology of a second aspiration showed, besides psammous bodies, some papillary components containing medium sized cells with rough nuclei, clear nucleoli and even some nucleic vacuoles. Based on fine needle biopsy, a papillary carcinoma of the thyroid was initially suspected.

c) Neck surgery

A left *cervicotomy* with total removal of a large, supra- and infraclavicular tumor measuring 7 × 5 cm in diameter, with a clear margin of healthy tissue around it, was performed on 1 July 1992. The tumor capsule was not incised or ruptured. There was no direct link to the thyroid. As the tumor was firmly adherent to the left jugular vein, the vein was resected en bloc with the tumor. The postoperative course was uncomplicated.

d) Histology

Pathologic macroscopic examination revealed a solitary tumor with a thin, but intact capsule of fibrous tissue. Cross sections revealed multiple haemorrhagic and serous cysts (±50% of the volume) being separated by yellow-whitish solid areas. *Microscopically*, the cystic spaces were lined by columnar epithelia bordering regular serous salivary gland parenchyma (approximately

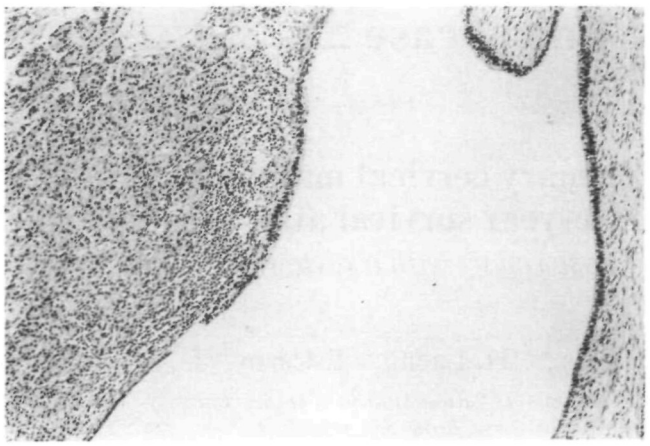

Figure 3 Histological section: Cyst lined by columnar epithelium bordering regular serous salivary gland parenchyma with loose lymphohistiocytic infiltrates (H & E, original magnification × 87).

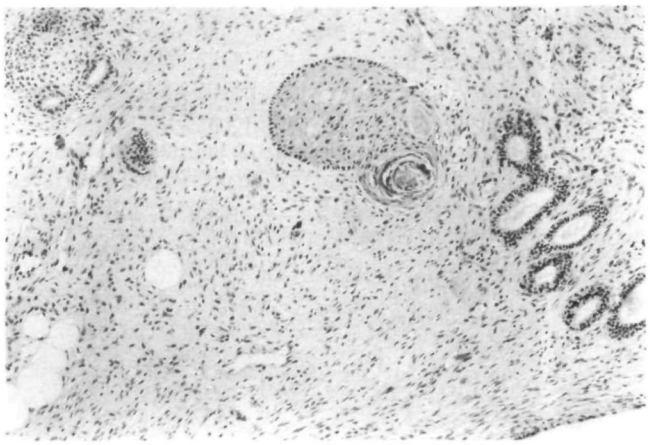

Figure 4. Histological duct structures, which may represent remnants of mature salivary glands. A solitary concentric focus of squamous epithelial metaplasia is present within abundant irregularly arranged fibrous stroma (H & E, original magnification × 87).

10% of the total volume) with patchy lymphocytic infiltrates (Figure 3). Thyroid parenchyma was not seen. The composition of the remainder of the tissue volume was heterogeneous. Intermingled with mature duct structures which might represent remnants of salivary gland ducts, concentric foci of squamous epithelial metaplasia in irregularly arranged loose fibrous stroma were present (Figure 4). Other mesenchymal counterparts (±30% of the volume) were made up of mature smooth muscle, but there was also hypercellular hyaline cartilage with slight irregularities in cellular size and shape, which suggested a grade 1 chondrosarcoma (Figure 5). In addition, tubular epithelial structures resembling embryonal carcinoma with occasional yolk sack-like projections were frequent. A few true rosettes, typical of neuroepithelial differentiation were present [6, 7, 9] (Figure 6). They were growing in an immature mesenchymal stroma, also displaying cytomorphologic atypia and mitotic activity (Figure 6), while additional features of so-called unfavorable histology such as rhabdomyosarcoma were absent. Scattered haemorrhagic necrosis was present in the malignant

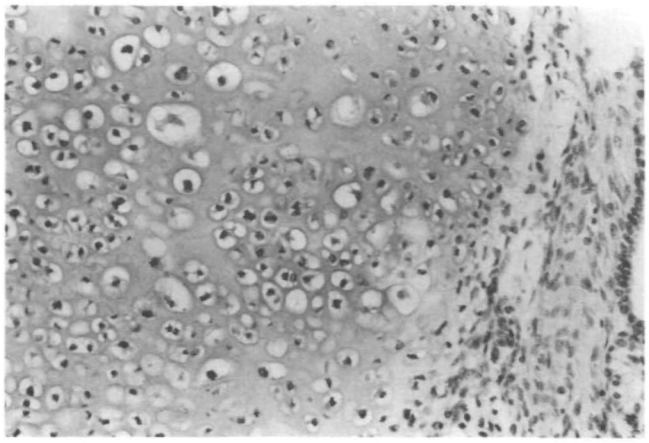

Figure 5. Histological section: Hypercellular mature hyaline cartilage with variation in cellular size and shape is suggestive of low-grade chondrosarcoma and is a frequent component of the mesenchymal counterpart of the tumor (H & E, original magnification × 175).

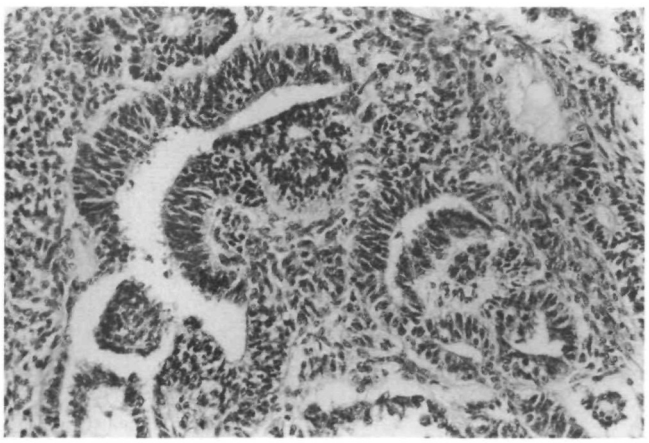

Figure 6. Histological section: Malignant epithelial counterpart of the tumor resembling embryonal carcinoma with yolk sack-like structures (arrow) and true rosettes (arrowhead) typical for neuroepithelial differentiation. The immature stroma displays mitotic activity (H & E, original magnification × 175).

components. A minimal local invasion of the intact fibrous capsule and of the sinuses of a small neighboring lymph node was seen. The conclusion was that this was a malignant teratoma (or teratocarcinoma) with components of both germ cell and of differentiated mesenchymal tumor. According to the WHO classification, this entity with two tumor components is distinct from an extragonadal germ cell tumor of mixed type, that lacks differentiated (e.g., non-germ cell) tissue.

e) Postoperative investigations

In order to differentiate between a primary or a metastatic teratoma to the neck, further diagnostic procedures were performed. The *laboratory findings* of: *alphafoetoprotein, lactate dehydrogenase, human chorionic gonadotropin, human thyreoglobuline* were normal. A clinical examination by an urologist was normal. A *computerized tomography of the pelvis* and of the *abdomen* and an *ultrasonography of the testicles* showed no pathologic tumor masses. A *computerized tomography of the thorax* again showed the known swelling of the 10th lateral left rib, compatible with an enchondroma. A *bone scintigraphy with 99mTc-MDP* showed no sign of skeletal metastases and in particular, the scintigraphic findings within the swollen 10th left rib were normal. During the next four months, the patient refused surgical resection of the rib mass. Finally, on 6 November 1992, a partial *excision of the 10th left rib* with total tumor resection was performed. Histology revealed a grade 1–2 chondrosarcoma, which probably represented a rib metastasis of the known malignant teratoma, although another primary tumor could not be formally excluded.

f) Adjuvant chemotherapy and outcome

Immediately after neck surgery, the medical intention was to treat with adjuvant chemotherapy. This is why, prior to the surgical rib resection, which was initially refused by the patient, two cycles of classical BEP *chemotherapy* (bleomycin: absolute doses 30 mg days 1, 8 and 15; etoposide: 100 mg/m^2 over one hour from day 1–5 included, and cisplatin: 20 mg/m^2 intravenously as a 30 minutes infusion from day 1–5 included [5]) were administered (September to October 1992). The chemotherapy was intended to be an adjuvant treatment, though the subsequent finding of a rib chondrosarcoma led to the conclusion that chemotherapy had instead been given for metastatic disease. Chemotherapy was well tolerated. Since then, the patient has remained free of symptoms and in good condition for more than five years (five years and 10 months at the time of this report) since resection of the primary neck tumor. Regular clinical, radiologic and blood serum tests have not indicated any sign of tumor recurrence.

We have given two adjuvant cycles of chemotherapy according to our policy in patients with testicular stage I germ cell tumors with elements of embryonal histology, where a relapse rate of 30% is to be expected. This policy was shown to reduce the relapse rate from 30% to 5% in this group of patients [44]. Since the rib lesion was scintigraphically negative and initially judged to be benign, additional chemotherapy was not thought to be necessary. Only after excision did it become clear that it was more likely a metastasis, possibly cleared of embryonal elements due to previous chemotherapy. Also the possibility of a synchronous multifocal teratocarcinoma can formally not be ruled out.

Discussion

a) Classification

Teratomas are tumors composed of several different tissues foreign to the anatomic site of origin in which they arise. They consist of all three germ layers: ectoderm, mesoderm and endoderm. A fine capsule is the rule. Microscopic examination reveals a variety of tissues

Table 1. Adult cervical malignant teratoma (locations at the skull base and along the cervical spine omitted): Chronological literature review of 24 published patients, including ours.

Publication		Preoperative data							Treatment			Clinical outcome				
Reference	Country	Sex	Age (y)	Tumor ∅ (cm)	Location	Path.	Symptom duration	Imaging	Surgery	Radiation therapy (Gy)	Chemo-therapy	Local recur-rence	Distant metastases	Survival (months)	Cause of death	Remarks
1 Lurje, 1908 [11]	CH	F	52	Massive	Thyroid ?	Mal	?	No	No	No	No	Yes	Lu: sarcoma	1	Metastatic tumor	Diagnosis at autopsy
2 Waechter, 1909 [12]	Germany	M	71	?	Thyroid	Mal	?	No	?	?	?	?	?	?	No follow-up	Quoted by [12]
3 Fritzsche, 1920 [12]	CH	F	41	10	Thyroid	Mal	5 weeks	No	Yes	No	No	Yes	Med, bo, lu	1	Asphyxia	
4 Buckwalter, 1954 [13]	USA, w	F	23	?	Thyroid	Mal	6 months	X, I	Yes	Neck, med	No	Yes (sarcoma)	Med, lu, pleu	9	Sudden, jug. vein infiltr	No autopsy
5 Kemp, 1967 [19]	Autralia, w	F	85	5	E-thyroid	Mal	50 years	X	Yes	No	No	No	No	Alive > 7	Incompl. follow-up	
6 Hajdu, 1967 [21]	USA, w	F	68	17	Thyroid	Mal	8 months	Yes	Yes	Recurr.	No	Yes	Med, lu, pleu	9	Sudden asphyxia	Autopsy [cf 36]
7 Pilheu, 1969 [22]	Argentine	M	27	14	Thyroid	Mal	12 months	NR	Yes	No	No	No	Lu	?	No follow-up	
8 O'Higgins, 1975 [23]	Irak	M	23	8	Thyroid	Mal	24 months	I	Yes	Yes	Yes	Cerv. ln	No	10	Local recurrence	
9 Colton, 1978 [24]	USA	M	77	10	Neck	Mal	7 months	X	2×	No	No	Yes	No	?	No follow-up	
10 Kimler, 1978 [25]	USA, b	F	37	10.5	Thyroid	Mal	3 months	US, X, NB	Yes	Yes	Yes	Yes	No	6	Gastric aspiration	Pregnant, child well
11 Dhellemes I, 1979 [27]	France	F	28	?	Thyroid	Mal	3 months	NR	Yes	Recurr.	Yes	Yes	Lu	14	Carotid rupture	
12 Dhellemes II, 1979 [27]	France	F	20	Small	Thyroid	Mal	4 months	NR	Yes	No	Yes	Yes	No	22	NR	
13 Murao, 1979 [28]	Japan	F	19	5	Thyroid	Unc	4 years	I, X	Yes	Yes	No	No	No	Alive > 8	Incompl. follow-up	Path. descrip-tion incompl.
14 Przystasz, 1979 [2]	Poland	F	48	6	Thyroid	Mal	4 months	X, I	Yes	46	Yes	No	No	?	No follow-up	
15 Trotoux, 1979 [29]	France	F	28	6	Thyroid	Mal	4 months	NR	Yes	Yes	Yes	Yes	lu	11	NR	
16 Tobey, 1980 [30]	USA	M	51	6	Neck	Mal	5 weeks	X, Ba, Tc, IVP	Yes	No	Cy, C, B, Ac	Cerv. ln	Med, retrocrural	Alive > 9	Incompl. follow-up	
17 Ceriani, 1984 [31]	Italy	M	19	8	Thyroid	Mal	3 months	X, Tc	2×	Recurr	No	3×	Med, lu, pleu, skin	9	Asphyxia	Autopsy
18 Kier, 1985 [32]	USA	M	27	3	Thyroid	Mal	12 months	CT	Yes	Yes	NR	NR	NR	NR	No follow-up	
19 Buckley, 1986 [33]	USA, w	M	27	20	Thyroid	Mal	12 months	FN, X, CT	Yes	Yes	Cy, A, E, C	No	Lu, meningia	2	NR	Local fistula, thyroid or.
20 Mochizuki, 1986 [34]	Japan	F	26	5	Neck	Mal	Asymptom.	X, I	Yes	No	No	No	No	Alive > 16	Incompl follow-up	Pat unnoticed, thyroid or.
21 Jordan, 1988 [36]	USA	F	68	17	NR	Mal	8 months	NR	Yes	Recurr.	No	Yes	Med, lu, pleu	8	Sudden asphyxia	Autopsy [cf 21]
22 Kahle, 1990 [37]	Germany	F	45	5.5	Thyroid	Mal	Immediate	US, FN	2×	Yes	Vb, C, B	Yes	Lu, li, bo	11	Cachexia, cardiac failure	Strumectomy < 2 years: ben.
23 Cain, 1991 [38]	Australia	M	38	2	St.cl.mast.L	Mal	2 weeks	Ba, CT, US	Yes	No	E, C, B	Yes, 5 cm	Lu, brain	3	Cachexia	hCG mediated hyperthyr
24 Als, 1998 [this public.]	CH	M	33	7	Neck	Mal	1 month	TcMIbl, US, X, CT	Yes	No	C, E, B, adj	No	bo (sarcoma)	well > 70	Probably cured	Adjuvant chemotherapy

Chemotherapy is detailed where known; in all patients (except ours), it was administered in the presence of recurrence or of metastases. Note that the case reported by Hajdu in 1967 [8] was later reported again by Jordan in 1988 [36]
Abbreviations: w white; b black; F – female; M male; y – years, e – extra, NR – not reported; St.cl.mast L – left sterno-cleido-mastoid muscle; path – pathology, Mal – malignant; unc – uncertain, ben – benign, CT – computerized tomography; US ultrasound; Tl – 201-Tl thyroid scan; MIBI – 99mTc-methoxyisobutylisonitrile thyroid scan, Tc – 99mTc pertechnetate thyroid scan, I – radioiodide thyroid scan; X – X-ray; Ba – barium swallow; NB – needle biopsy; FN fine needle aspiration cytology; IVP – intravenous pyelography; ln – lymph nodes, cerv – cervical; med – mediastinum, lu – lung; bo – bone; pleu – pleura; li – liver; or – origine; recurr – recurrence; adj – adjuvant; A adriamycine; Ac actinomycine D; B – bleomycine; C – cysplatine, Cy – cyclophosphamide; E – vepesid; V – vincristine, Vb – vinblastine; incompl – incomplete

Table 2. Adult cervical benign teratoma (locations at the skull base and along the cervical spine omitted): Chronological literature review of 11 published cases.

Publication		Preoperative data							Treatment			Clinical outcome				
Reference	Country	Sex	Age (y)	Tumor Ø (cm)	Location	Path.	Symptom duration	Imaging	Surgery	Radiation therapy (Gy)	Chemo-therapy	Local recur-rence	Distant metas-tases	Survival (months)	Cause of death	Remarks
1 Marescot, 1945 [3]	Spain	F	36	Massive	Neck	Ben.	21 years	No	Yes	No	No	NR	NR	?	No follow-up	
2 Cavallero, 1954 [14]	Italy	M	46	Nutsize	Thyroid	Ben.	20 years	NR	Yes	No	No	> 22 y	No	> 22 y	Cured	Structectomy 2 years before
3 Bale, 1959 [15]	USA	?	?	?	Neck	Ben.	?	X	Yes	No	No	Yes	NR	NR	NR	
4 Keynes, 1959 [16]	GB, w	M	24	5	Thyroid	Ben.	24 years	NR	Yes	No	No	Yes	NR	Well > 15	Incompl. follow-up	Since childhood, recurr.
5 Notcboom, 1959 [17]	USA	M	19	4	Thyroid	Ben.	NR	X	Yes	No	No	NR	NR	NR	Incompl. follow-up	
6 Prüfer, 1965 [18]	Germany	F	18	7	Thyroid	Ben	10 months	X	Yes	No	No	No	No	Well > 60	Probably cured	3 healthy babies born
7 Stone, 1967 [20]	USA, b	F	20	6	Thyroid	Ben.	22 months	X, Ba	Yes	No	No	No	No	Alive > 135	Probably cured	Path. description incompl.
8 Woods, 1978 [26]	USA, w	F	40	?	Neck	Ben.	18 months	Tc, X, Ba	Yes	No	No	No	No	Alive > 12	Incompl. follow-up	
9 Cannon, 1987 [35]	USA	F	32	32	Larynx	Ben.	3 months	NR	Yes	No	No	NR	NR	Alive > 60	Probably cured	Hoarseness, larynx
10 Sawafuji, 1993 [1]	Japan	M	21	10	Neck	Ben.	3 months	X, CT, MRI, Tc, Tl	Yes	No	No	No	No	Well > 19	Incompl. follow-up	201-Tl scan positive
11 Kuhel, 1996 [39]	USA, w	F	32	3	Thyroid	Ben.	1 month	CT, MR	No	No	No	No	No	Well > 22	Incompl. follow-up	CT/MR; fat areas

Abbreviations: see Table 1.

with a wide range of cellular differentiation and variable degrees of maturation. Immature components corresponding most often to germ-cell tumors seem to be of lesser prognostic significance in infants [6] but represent a poor prognosis in adults [7].

Since the first report of a neck teratoma by Hess in 1854 [10], about 230 cases have been reported in all ages, of which 34 were in adults (Tables 1 and 2) [1–3, 11–39]. 'Cervical teratoma', the term used nowadays, generally incorporates lesions arising in the anterior and posterior triangles of the neck. Cervical teratomas are rare and represent only 2%–9% of all reported teratoma cases [40]. The term does not include teratomas of the skull base or along the cervical spine [7]. The rare event of a cervical teratoma in an adult should be viewed in a more general context. By far, most teratomas occur in infants before the age of one year and are benign, many of them are present at birth [6, 8]. In adults, teratomas are very rare; 70% are malignant. Teratomas most often occur in para-axial or mid-line location from the brain to the sacral area as well as in the gonads. Table 3 illustrates that the primary tumor location shows different decreasing frequencies in children and adults [7, 9, 45].

The former clinical distinction between a thyroid or neck origin of a teratoma, – according to: 1) origin of feeding vessels, 2) direct continuity between tumor and gland, 3) localization of tumor within the gland, 4) entire absence of the gland, – has no practical significance because there are no differences in prognosis or treatment [36]. Moreover, these criteria may be misleading because it is often impossible to determine the vascular connections of the tumor. Thyroid parenchyma within the tumor may represent remaining thyroid from which the teratoma arose, or well differentiated elements arising in the teratoma itself, or even thyroid tissue secondarily invaded by the teratoma.

b) Clinical presentation of cervical teratoma in adults

Out of the 35 reported cases (our case included) of cervical teratoma in adults, covering various ethnic origins (Tables 1 and 2), 32.5% were benign and 68.5% (one uncertain case included) were malignant. The tumor diameter at presentation varied from 3 cm to mas-

Table 3. Decreasing frequencies of the locations of a primary teratoma in infants and children as compared to adults [7, 9, 40, 48].

Infants and children		Adults
Sacrococcygeal region	40%	Ovary
Ovary	37%	Testes
Head and neck	5.5%	Sacrococcygeal region
Retroperitoneum	5%	Mediastinum
Mediastinum	4%	Retroperitoneum
Brain and spinal cord	3.5%	Central nervous system
Testes	3%	Liver
Liver	1%	Nasal sinuses
Abdominal wall, para-umbilical	< 1%	Neck
Scapula (back)	< 1%	

sive tumors of more than 30 cm. The median symptom duration before operation was much longer with benign than with malignant teratomas. Typically, rapid progression started after a long dormant phase. Metastases and local recurrences were often sarcomatous.

The median age of the 35 patients was 32 years (range 19–85 years). In 90% of benign teratomas and 58% of malignant teratomas, the diagnosis was made before the age of 40 years. The higher the age at diagnosis, the more likely was the malignant nature of the tumor: the median age in case of benign disease was 24 years, in case of malignant disease 35 years. Mochizuki [34] had postulated that cervical teratoma in adults undergoes a rapid change in biological behavior from benign to malignant above the age of 20 years, as the youngest malignant case was seen in a 19-years-old female [28].

c) Imaging

Because most tumors were clinically suspected to be related to the thyroid, previous imaging of cervical teratomas was reported as thyroid scintigraphy (99m-Tc pertechnetate or radioactive iodine), tumor scintigraphy with 201-Tl or 99mTc-MIBI, ultrasound or CT (Tables 1 and 2). In case of papillary thyroid carcinoma, tumor scintigraphy with 99mTc-MIBI has been reported positive [4, 41, 42]. However, it was negative in our patient.

d) Treatment and prognosis

The prognosis of benign teratoma is generally good (Table 2). But the prognosis of malignant teratoma is sombre (Table 1): none of the patients with prolonged follow-up was cured, despite various treatment protocols. Thus, the maximal survival of the malignant cervical teratoma reported to date was 22 months (median nine months, average eight months, Table 1). External radiotherapy since the 1950's and, since the late 1970's, chemotherapy, relying today mostly on cisplatin-containing regimens [43], have been administered after surgical excision. These treatments were always applied late in the course of the disease, when there were large tumor burdens, and their effect was only palliative. For other extragonadal teratocarcinomas such as those of the anterior mediastinum, the prognosis is inferior to the one of testicular teratocarcinomas with longterm survivals of 40%–50%, where as such tumors arising from the ovaries (non-dysgerminomatous germ-cell tumors) have an excellent survival after intensive chemotherapy, comparably to testicular teratocarcinoma [46, 47].

In contrast, our patient with a cervical malignant teratoma with components of germ cell and of mesenchymal tumors was given BEP-chemotherapy [5] with adjuvant intent in the postoperative course based on clinical experience with germ cell tumors in general. In a British study of clinical stage I patients with non-seminomatous germ cell tumors of the testicle, the pre-study relapse rate was 30%. This was reduced to 5% by a treatment policy of two courses of adjuvant bleomycin, etoposide and cisplatin (BEP). This 5% relapse rate compared favorably with that of 18.5% in clinical stage I patients treated by surgery alone [44]. Therefore, adjuvant BEP-chemotherapy in the postoperative course in general is effective in preventing relapse in patients with testicular germ cell tumors in general and, hopefully with malignant teratoma (with germ-cell components).

As is the case with chemotherapy, *prognosic factors* for malignant teratoma are deduced from those of germ cell tumors in general. For testicular germ-cell cancer, the extent of disease and serum tumor-marker concentrations were identified as independent predictors of prognosis, and previously untreated patients with metastatic disease were subdivided into good-risk and poor-risk groups [50, 51]. This staging system was developed because of the high cure rate and substantial toxicity of combination chemotherapy and a need to identify patients more likely ('good-risk') and less likely ('poor-risk') to be cured by standard chemotherapy. In another British multicenter study on non-seminomatous germ-cell tumors of the testicle, the only significant independent prognostic factors were based on histology and lymphatic invasion [48]. The relapse rates ranged from six out of 50 cases (12%) in teratocarcinoma patients without lymphatic invasion to seven out of nine cases (78%) of embryonal carcinoma patients with lymphatic invasion [49]. By analogy, our patient belonged to the high-risk category for relapse because of embryonal components in the primary tumor with minimal lymphatic invasion and because of the bone metastasis (excised chondrosarcoma in the 10th left rib). This metastasis was already present at the time of the neck operation, albeit not yet pathologically proven because of the patient's initial refusal of rib surgery. Nevertheless, the patient has remained tumor free for nearly six years after neck surgery. The en bloc surgical removal of the tumor without rupture of the capsule, followed by BEP-chemotherapy and complete removal of the solitary metastasis, appear to have eradicated the tumor. Five-year progression-free survival rates between 66% and 78% according to histological findings of postchemotherapy resections of residual masses from metastatic non-seminomatous testicular germ-cell tumors retrospectively confirm the rightness of our therapeutic attitude. It especially confirms how essential the primary en bloc surgical removal of the tumor is [46]. This is amplified since, as also shown in mediastinal non-seminomatous germ-cell tumors, only patients who achieve disease-free status after cisplatin-based combination chemotherapy are likely to be cured [47].

Of the previously reported patients in the literature (Tables 1 and 2), [1–3, 11–39], our patient was the only one, where there was a decision to treat with adjuvant chemotherapy. This meant that there was no delay in the patient receiving chemotherapy for what turned out to be metastatic disease. Compared to the sombre prognosis of cervical malignant teratoma in adults (Table 1), the good clinical five-year outcome of our patient suggests

that the use of 'adjuvant' BEP-chemotherapy, – as classically used with germ cell tumors, – immediately after the neck resection and independently of the pathologic nature of the rib tumor, was appropriate. We suggest that a policy of adjuvant chemotherapy should be examined in other non-gonadal malignant teratoma patients in the future, in order to gain prospective evidence of its possible beneficial influence on prognosis.

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